Aberrant Femoral Torsion Presenting with Frog-leg Squatting Mimicking Gluteal Muscle Contracture

Chia-Ling Chiang MD, Meng-Yuan Tsai MD,
Wei-Ning Chang MD, Clement Kuen-Huang Chen MD

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Abstract

Background Patients with frog-leg squatting have restricted internal rotation and adduction of the affected hips during sitting or squatting. In the surgical literature, the cause generally has been presumed to arise from and be pathognomonic for gluteal muscle contracture. However, we have encountered patients with frog-leg squatting but without gluteal muscle contracture.

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Each author certifies that Kaohsiung Veterans General Hospital approved the human protocol for this investigation and that all investigations were conducted in conformity with ethical principles of research.

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C.-L. Chiang, M.-Y. Tsai, C. K.-H. Chen
Department of Radiology, Kaohsiung Veterans
General Hospital, Kaohsiung, Taiwan

W.-N. Chang
Department of Orthopaedics, Kaohsiung Veterans
General Hospital, Kaohsiung, Taiwan

C. K.-H. Chen (✉)
Department of Medical Imaging, Chi Mei Medical
Center, Number 901, Zhonghua Road, Yongkang District,
Tainan City 71004, Taiwan
e-mail: ckhc821@gmail.com

C. K.-H. Chen
National Yang-Ming University School of Medicine,
Taipei, Taiwan

Questions/purposes We therefore raised the following questions: What are the imaging features of patients with frog-leg squatting? Do conditions other than gluteal muscle contracture manifest frog-leg squatting?

Patients and Methods We retrospectively reviewed the MR images of 67 patients presenting with frog-leg squatting from April 1998 to July 2010. There were four females and 63 males; their mean age was 22.2 years (range, 4–50 years). During MRI readout, we observed aberrant axes of some femoral necks and obtained additional CT to measure femoral torsion angles in 59 of the 67 patients.

Results MR images of 27 (40%) patients had signs of gluteal muscle contracture. Twenty-two (33%) patients (40 femora) had aberrant femoral torsion, including diminished anteversion (range, 6°–0°; average, 3.9°) in 11 femora of eight patients and femoral retroversion (range, < 0° to –31°, average, –7.5°) in 29 femora of 17 patients. The remaining 18 (27%) patients did not have gluteal muscle contracture or aberrant femoral torsion. The observation of aberrant femoral torsion was not anticipated before imaging studies.

Conclusions In addition to gluteal muscle contracture, aberrant femoral torsion can be a cause of frog-leg squatting.
Level of Evidence Level II, diagnostic study. See the guidelines for Authors for a complete description of levels of evidence.

Introduction

Frog-leg squatting is used to describe patients who are unable to sit or squat with their knees close together (Fig. 1). Instead, they typically present with hip abduction and external rotation when crouching [2, 6]. Individuals with severe cases cannot sit or squat without abduction of

Fig. 1A–D A comparison of the squatting position in a healthy adult and a patient with GMC is shown. **(A)** A healthy adult can easily squat with both knees close together. **(B)** A 50-year-old man with bilateral asymmetric GMC (left side more severe than right side) has

limited adduction of both hips during sitting and squatting. **(C)** A healthy adult can easily squat with both knees wide open. **(D)** The patient cannot squat completely even with his knees wide open owing to severe GMC and the resultant limited flexion.

the thighs, giving the appearance of the limb position of a frog. In the literature, frog-leg squatting has been considered the result of and pathognomonic for gluteal muscle contracture (GMC) [2, 6]. Most of the reported patients are school-aged children (6–18 years old), and the lesions are often secondary to repeated intramuscular injections in the buttocks [2, 6, 8, 9, 14] and a constitutional fibrogenic diathesis in many patients [2, 7]. GMC has been used as one of the criteria for exemption from military service in Taiwan. However, for military certification, the orthopaedists in our hospital expect some objective image evidence of GMC in addition to the physical examination. Apart from confirming the clinical diagnosis, MRI can contribute to thorough evaluation of the disease extent and severity and assist in preoperative planning [2]. Therefore, we routinely obtained MRI of the pelvis for these patients.

When we instituted routine MRI we focused on determining whether the images contained evidence of GMC. However, we observed some patients with normal gluteal muscle but with femoral retroversion (a major cause of femoroacetabular impingement [FAI]) [12]. This prompted us to ask whether disorders other than GMC could be associated with frog-leg squatting. Therefore, we began observing the femoral neck axes on the axial MR images of the pelvis in patients with frog-leg squatting and began to routinely obtain supplemental CT to detect femoral torsion. The femoral neck axis of a normal healthy adult leans not only medially but also relatively anterior to the transcondylar axis by 15°, which is defined as anteversion [5, 11]. In patients with aberrant femoral torsion, the femoral neck axis has a smaller than normal anteversion angle [13] or even retroversion when the femoral neck axis leans relatively posterior to the transcondylar axis [3, 4].

We therefore asked: (1) what are the imaging features of frog-leg squatting, and (2) is there any condition other than GMC manifesting similar symptoms?

Patients and Methods

After obtaining institutional review board approval (VGHKS11-CT4-06), we retrospectively reviewed the MR and CT images of all 67 patients with a clinical diagnosis of GMC who were referred from the orthopaedic outpatient clinic of our hospital between April 1998 and July 2010. Of the 67 patients, 63 were males and four were females. Their mean age was 22.2 years (range, 4–50 years). All 67 patients underwent MRI, and supplemental CT torsion studies were performed in 59 (88%) patients.

Most of the patients had variable degrees with restriction of internal rotation and adduction of their hips. Physical examinations included active and passive squatting tests. In an active squatting test, the examiner asks the patient to squat by himself or herself with both knees close together. The test was positive if the patient was unable to accomplish the posture and ultimately fell backward. In a passive squatting test, the patient lay on the examination table and the examiner brought the flexed knee close to the patient's chest with provocation of pain on flexion and internal rotation. The test was positive if there was resistance with hip flexion less than 90°, and the patient usually was unable to tolerate the pain [2].

MRI of the pelvis focusing on both hips and buttocks was performed with a 1.5-T scanner (Signa; GE Healthcare, Waukesha, WI, USA), with a standard torso coil and the patient lying in the supine position. The pulse sequences included multiplanar T1-weighted and fat-saturated proton-density and T2-weighted images [2]. MRI of a GMC typically shows primary and secondary features [2] (Table 1). Primary features include gluteal muscle atrophy or intramuscular fibrotic cords. The fibrotic cords show low signal intensity on all sequences and were most obvious on fat-saturated images (Fig. 2). Thickened tendon and fibrotic cords produce secondary features including

medial retraction of the affected gluteus maximus muscle, medial posterior displacement of the iliotibial tract behind the greater trochanter, depressed groove of the muscle-tendon junction, and external rotation of the proximal femur [2] (Fig. 3).

The CT torsion study was performed with a 16-MDCT unit (Somatom Sensation 16; Siemens Medical Solutions, Malvern, PA, USA). Two of us (MYT, CKHC) calculated femoral version in the following way [3–5]: we superimposed an image through the axis of the longest femoral neck and a transcondylar image of the distal femur to obtain the two required axes. A femoral neck axis was drawn as the midline bisecting the femoral neck on an axial CT slice with the longest femoral neck. A transcondylar

femoral axis was drawn as a straight line tangential to the posterior cortices of both medial and lateral condyles of the distal femur. We obtained the femoral neck and transcondylar femoral axes from CT images of the two levels and measured the angular difference between the two axes (Fig. 4). Version of the proximal femur refers to the relationship of the axis of the femoral neck to the transcondylar axis of the distal femur. Femoral anteversion refers to the condition where the femoral neck axis leans anterior to the transcondylar femoral axis. At birth, neonates have an average of 40° to 45° femoral anteversion [11, 13], whereas by age 8 years, average anteversion decreases to the typical adult value of 15° [5, 11, 13]. If the anteversion angle is smaller than the average value for patients of the same age, it is termed diminished anteversion [13]. Femoral retroversion refers to the condition where the femoral neck axis is oriented posterior to the transcondylar axis, thus positioning the femoral neck and head posterior to the coronal plane of the femur. In this article, aberrant femoral torsion refers to either diminished anteversion or femoral retroversion but not excessive femoral anteversion. The measurement method for the femoral neck axis is widely accepted [3, 4], although intraobserver/interobserver variance has been reported [5]. The major cause of variability is the image selection for femoral neck axis measurement. We chose the axial image with the longest view of the femoral neck and reached a consensus for every case.

Table 1. Causes of frog-leg squatting

Causes	Number of patients
Gluteal muscle contracture	27/67 (40%)
Primary MRI features	
Muscle atrophy	27 (49)*
Intramuscular fibrotic cord	25 (44)*
Secondary MRI features	
Medial posterior displacement of iliotibial tract	11 (17)*
Medial retraction of gluteus maximus muscle	10 (15)*
Depressed groove at muscle-tendon junction	10 (15)*
External rotation of proximal femur	9 (13)*
Aberrant femoral torsion	22/67 (33%)
CT torsion study identified	
Diminished anteversion	8 (11) [†]
Femoral retroversion	17 (29) ^{†,‡}
Normal gluteal muscle and normal femoral version	18/67 (27%)

* Values are expressed as number of patients (number of buttocks);

[†]values are expressed as number of patients (number of femoral necks); [‡]three patients had diminished anteversion on one side and femoral retroversion on the other side.

Results

Eighteen of 67 (27%) patients had normal gluteal muscles and normal femoral anteversion, 27 (40%) patients had typical findings of GMC, and the remaining 22 (33%) patients had aberrant femoral torsion (Table 1). Forty-nine buttocks of the 27 patients with typical GMC showed primary features.

Fig. 2A–B A 50-year-old man with long-term frog-leg squatting has bilateral GMC that is more severe on the right side. (A) Coronal T1-weighted and (B) fat-suppressed T2-weighted images show bilateral atrophic gluteus maximus muscles with fatty replacement

(black arrows) and hypointense intramuscular fibrotic bands (dotted arrows) extending downward and outward obliquely. (C) An axial T1-weighted image shows bilateral atrophic gluteus maximus muscles (black arrows) and an intramuscular fibrotic band (dotted arrow).

Fig. 3A–C A 25-year-old man has bilateral GMC, more severe on the right side. **(A)** An oblique coronal T1-weighted image shows an atrophic gluteus maximus muscle with fatty replacement (arrows) and an intramuscular fibrotic band (arrowheads). **(B)** An axial T1-weighted image shows bilateral gluteal muscle atrophy (black arrows), more severe on the right side, with an intramuscular fibrotic band (dotted arrows), medial retraction of the gluteal muscle, and depressed muscle-tendon junction. **(C)** An axial T1-weighted image at a more inferior level than Illustration B shows bilateral gluteal muscle atrophy (black arrows), medial posterior displacement of the bilateral iliotibial bands (white arrows), and external rotation of bilateral proximal femurs (double-headed arrows).

Among the 59 patients receiving CT torsion studies, 40 femora from 22 patients had aberrant femoral torsion, including diminished anteversion (range, 6° – 0° ; average, 3.9°) in 11 femora of eight patients and femoral retroversion (range, $<0^{\circ}$ to -31° ; average, -7.5°) in 29 femora of

17 patients. Three patients had diminished anteversion on one side and femoral retroversion on the other side.

Discussion

Frog-leg squatting is used to describe a position observed in patients who have difficulty in sitting or squatting with their knees close together and who have to open their thighs wide during sitting or squatting, thus resembling frog legs. The clinical symptom of frog-leg squatting has long been regarded as a characteristic manifestation of or even pathognomic of GMC based on reported cases of frog-leg squatting in school-aged children (6–18 years) [2]. We investigated the imaging features of frog-leg squatting and whether there is any disorder other than GMC also manifesting similar symptoms?

There are some limitations to our study. First, for the 18 patients with normal MRI and CT, the possibilities of malingering or other unknown causes require further investigation. However, in this retrospective descriptive study we had not begun to survey other causes in patients with frog-leg squatting at the time these patients were treated. Second, to the best of our knowledge, the incidence of aberrant femoral torsion is not well established in the general population. Therefore, although we found a high percentage of aberrant femoral torsion in the patients with frog-leg squatting, it is difficult to confirm either the statistical significance or clinical importance. Third, we encountered a few cases with variable measurements. We used the widely accepted method for femoral torsion measurement [3, 4], and carefully selected the axial image with the longest view of the femoral neck. Through proper image selection and consensus discussion, the variability can be decreased. Fourth, our hospital is a referral medical center, and most patients were referred here for second authentication and certification only. We had little information regarding the treatment or followup for these patients.

GMC is widely reported in Asian countries, with a reported incidence of 1% to 2.5% in children in China [6]. Numerous authors believe GMC is secondary to repeated intramuscular injections, regardless of the type of medication injected into the buttocks [2, 6, 8, 9, 14] and/or a constitutional fibrogenic diathesis [2, 7]. GMC is characterized by variable degrees of muscle atrophy and intramuscular fibrosis of the gluteus maximus muscle, which leads to restriction of internal rotation, adduction, and flexion of the affected hips. Patients with severe GMC usually seek medical help owing to waddling gait, squatting difficulty, or untraditional sitting or squatting posture. GMC is not uncommon in adolescents and young adults experiencing repeated intragluteal injections during

Fig. 4A–D (A) Several axial images were obtained from the femoral neck level and the femoral condylar levels and (B) then overlaid to measure the angle between the femoral neck axis (dotted line) and the transcondylar femoral axis (solid line). (C) The normal femoral neck

axis is anterior to the transcondylar femoral axis by 8° to 15° in the mature skeleton. (D) A CT torsion study shows the aberrant femoral neck axis posterior to the transcondylar femoral axis in a 22-year-old man with femoral retroversion.

childhood and has been regulated as one of the criteria for exemption from military service in Taiwan. Therefore, some young men of military service age (19–36 years) with variable degrees of difficulties in squatting seek medical evaluation to avoid recruitment. A positive active squatting test and a passive hip flexion angle less than 90° usually are present in patients with GMC. In healthy adults, the mean ($\pm$ SD) hip flexion angle is 121° ($\pm$ 13°) [2]. Although in the past, the diagnosis of GMC usually was derived from the history and physical examination, additional objective imaging studies are indicated not only for military certification but also for detailed evaluation of disease extent, degrees of muscle atrophy, and preoperative planning [2]. Operative release of fibrotic tissue with surgical division, resection [2, 14], or radiofrequency energy [6] effectively restores reasonably normal squatting in patients with severe GMC; nonoperative management such as passive stretching is recommended as a primary treatment for mild GMC [14].

Femoral retroversion usually occurs in adolescents or young adults, with complaints of anterior groin pain and difficulty in sitting or squatting with their knees close together or crossing their legs [10, 12], symptoms similar to the positive findings with the active squatting test in patients with frog-leg squatting. Patients with femoral retroversion also cannot tolerate the pain provoked by bringing the flexed knee to the chest [10], indicating positive anterior impingement [10], which is equivalent to the passive squatting test for frog-leg squatting. After reviewing the images and the literature, we suggest

diminished femoral anteversion or femoral retroversion results in a smaller anterior space between the anterior acetabular rim and contact surface of the femoral head-neck junction, restricting internal rotation, and a relative larger posterior space between the posterior acetabular rim and femur, allowing an increased capability of external rotation [1, 7, 12, 13]. A reduction in anterior joint clearance subsequently potentiates early apposition and repetitive impaction of the anterior acetabular rim and femoral head-neck junction during hip flexion, adduction, or internal rotation, leading to early osteoarthritis [1, 12, 13]. Twenty-two of our 67 patients (33%) with variable degrees of frog-leg squatting had aberrant femoral torsion with FAI rather than GMC. These patients were diagnosed with GMC clinically before MRI and CT and they could be regarded as malingerers after their MR images were negative for GMC. We observed no obvious degenerative change on the images of the patients with aberrant femoral torsion, which could be related to their generally young age; they were referred to our institution for certification and their only symptom was limited hip motion. GMC can lead to restriction of hip ROM and is a well known cause in Asian countries. In our clinical practice fibrosis and contractures are not uncommonly observed over the shoulder and buttock regions related to intramuscular injections and leading to deltoid or gluteal muscle contracture. Among 59 of our 67 (88%) patients who had CT and MRI, we found no causal link between GMC and femoral retroversion. Therefore, aberrant femoral torsion with FAI should be regarded as one of the major causes of frog-leg squatting.

For patients with GMC, MRI can provide an objective diagnosis and an accurate evaluation of extent and severity, and can assist in preoperative surgical planning [2]. The imaging features of frog-leg squatting include variable degrees of GMC, aberrant femoral torsion, and even negative findings. Frog-leg squatting in patients with negative MRI findings can be attributable to other unknown disorders or possibly the patient is a malingerer. We found, in addition to GMC, aberrant femoral torsion with FAI is associated with frog-leg squatting. The combination of MRI of the pelvis with a torsion study of the femur is helpful in detecting aberrant femoral torsion. Other occult causes of frog-leg squatting require further investigation.

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